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SCIENTIFIC LETTERS

Spontaneous hepatic portal venous gas in a patient with ulcerative colitis. A case report and a review

Gas venoso portal hepático espontáneo en un paciente con colitis ulcerosa. Reporte de un caso y revisión

Hepatic portal venous gas (HPVG) has been defined as the presence of gas/air in the portal venous system. It has been traditionally associated with ischemic bowel processes, and has been considered as an ominous finding with a mortality rate up to 75%.[1]

HPVG has been described in patients with inflammatory bowel disease (IBD), mostly in Crohn's Disease (CD).[2,3] We present a case of HPVG and a literature review of the reported cases of HPVG in ulcerative colitis (UC).

A 45-year-old-woman was diagnosed with left-sided UC 15 years ago. She started treatment with azathioprine after a steroid-dependent flare-up in 2010, adding infliximab in 2012 due to the persistence of UC activity.

In October 2014, the patient was admitted in our emergency care unit presenting acute severe abdominal pain referred to epigastrium, nausea, tachycardia and fever (39.3 °C). She did not report evidence of clinical UC activity in recent months. In serial blood samples an increase in inflammatory markers (C-reactive protein [CRP], up to 63 mg/L from normal values within the first 24 h) was observed. Simple chest and abdominal X-rays showed no relevant findings. An abdominal computed tomography (CT) was performed, and HPVG was observed in the left lobe of the liver (Fig. 1). Other intra-abdominal complications were ruled out and inflammatory changes in the rectum and sigma were observed, suggesting active UC. Immediately afterwards, a rectosigmoidoscopy was performed in which continuous mucosal damage with moderate inflammation up to 40 cm from the anal verge was identified.

The patient was admitted in our department for conservative management. Endovenous antibiotics (metronidazole plus ceftriaxone) and steroids (metyl-prednisolone 1 mg/kg daily) were initiated. Both azathioprine and infliximab treatment were discontinued.

The patient promptly reported feeling better and remained afebrile. Subsequent blood samples showed a progressive decrease of CRP (6.8 mg/L at discharge). Urine and blood cultures, as well as stool samples studies, were negative. At the 7th day of admission, a control CT-scan was performed, with complete HPVG resolution. Thereafter, endovenous steroids were changed for oral steroids. The patient remained in our unit for a total of 10 days and at the time she was discharged, antibiotics were stopped.

Liebman[1] reported 64 HPVG patients in which up to 72% cases were associated with bowel necrosis, with a global mortality rate of 75%, advocating for an aggressive management and urgent surgical exploration once this condition was diagnosed. More recently, an updated review of 182 patients with HPVG reported an overall mortality rate of 39%.[2] Other apparently more benign conditions, such as gastric ulcer, complications of endoscopic procedures and IBD, were associated with HPVG.[2] Some of these conditions reacted favorably to conservative management, suggesting that HPVG itself is not a predictor of poor outcome.[1,2,4,5]

Abdominal ultrasound and CT-scan are the radiological modalities used nowadays to diagnose HPVG.[3,5] The classical appearance of this entity is that of branching lucencies within 2 cm of the liver capsule, predominantly in the left lobe, which differs from biliary gas because the latter is associated with air within the central portion of the liver[1–5] (Fig. 1).

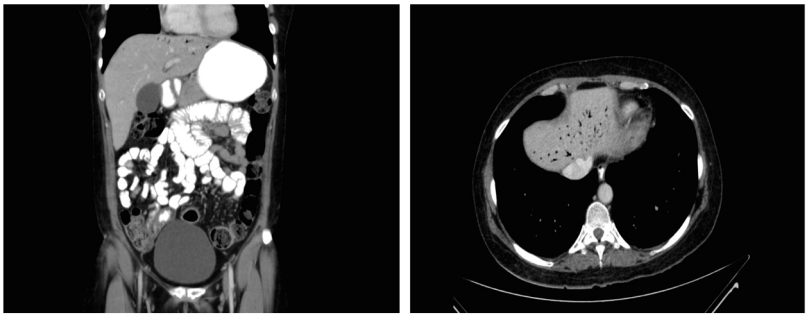

Figure 1 Computed tomography (CT)-scan images at diagnosis of HPVG. (a) Axial reconstruction. (b) Coronal reconstruction. Portal vein gas is predominantly located within the left hepatic lobe in both CT-scan images. Air bubbles appear to extend within 2 cm of the liver capsule.

Table 1 Cases of hepatic portal venous gas in ulcerative colitis.

Author	Gender	Age	UC extent	Years of UC	Prior procedure	Clinical features	Clinical UC activity	Diagnostic modality	Mucosal damage	Treatment	Outcome
Lazar et al.[6]	Male	48	Extensive	UK (1–3)	BE	Abdominal pain Fever Emesis	Flare	X-ray (plus perforation)	Yes	ATBs Sulfathalidine Prednisone	Recovery
Weinstein et al.[11]	Male	35	UK	6	BE	Malaise Chills Fever Tenderness	Remission	X-ray	Yes	ATBs	Recovery
Speer et al.[12]	Male	79	Left-sided[a]	10	None	Abdominal pain Low fever	Remission	X-ray	Yes	ATBs (as an outpatient)	Recovery
Kees et al.[9]	Female	44	Extensive	6	1-BE 2-BE	Low fever Mild TC	1-Flare 2-Remission	X-ray	1-Yes 2-Yes	Sulfasalazine, ATBs	1-Recovery 2-Recovery
Liebman et al.[1]	Male	23	UK	UK	BE	Lightheaded Shaking chill	Remission	X-ray	Yes	ATBs	Recovery
Christensen et al.[8]	Male	71	Left-sided[a]	5	BE	Mild stomach ache	Remission	X-ray	No	None	Recovery
Haber[13]	Female	45	Left-sided	4	RSC	Fever	Flare	X-ray	Yes	ATBs NG aspiration	Recovery
Birnberg et al.[14]	Female	24	UK	7	BE	Low fever	Remission	X-ray	Yes	ATBs	Recovery
Moss et al.[15]	Male	39	Extensive	<1	BE	Tenderness	Flare	X-Ray (plus perforation)	Yes	ATBs Surgery (subtotal colectomy)	Recovery
See et al.[16]	Female	53	UK	UK	Total colectomy due to fulminant colitis	Multiple organ failure	–	CT-scan	–	ATBs	Death
Bull et al.[7]	Female	60	Extensive	3	BE	None	Flare	X-ray	Yes	None	Recovery[b]
Paran et al.[10]	Male	51	UK	UK	None	Abdominal pain Bloodless diarrhea	Flare	CT-scan	Yes	Parenteral Nutrition ATBs	Recovery
Shah et al.[17]	Male	22	Extensive	UK	Total colectomy due to refractory UC	Abdominal pain Tachycardia Hypotension	–	CT-scan	-	Parenteral Nutrition ATBs	Recovery
Shinagawa et al.[18]	Male	18	Extensive	<1	Colonoscopy	Fever	Flare	CT-scan	Yes	ATBs	Recovery
Bamba et al.[19]	Male	54	Extensive	2.5	Colonoscopy	None	Flare	X-ray	Yes	Food deprivation	Recovery
Tanaka et al.[20]	Female	87	UK	22	None	Diarrhea Abdomnial pain Vomiting	Flare	CT-scan	Yes	ATBs	Recovery

Table 1 (*Continued*)

Author	Gender	Age	UC extent	Years of UC	Prior procedure	Clinical features	Clinical UC activity	Diagnostic modality	Mucosal damage	Treatment	Outcome
Fukita et al.[21]	Male	40	UK	9	None	Hematochezia Abdominal pain Feber	Flare	CT-scan	Yes	ATBs	Recovery
Present case	Female	45	Left-sided	15	None	Abdominal pain Fever	Remission	CT-scan	Yes	ATBs Prednisolone	Recovery

ATBs, Antibiotics; BE, Barium Enema; CT-scan, Computer Tomography-scan; NG, Naso-gastric; RSC, Rectoscopy; TC, Tachycardia; UC, Ulcerative Colitis; UK, Unknown, X-ray: plain film abdominal radiography.

[a] Minimum known extension.

[b] Panproctocolectomy due to UC flare nine days after admission.

The pathogenesis of HPVG could be explained as follows: (1) the passage of intraluminal air or gas-forming bacteria into the portomesenteric venous system due to mucosal disruption (2) increased bowel pressure that leads to bowel distension with mucosal disruption, described mainly in iatrogenic procedures such as colonoscopies and barium enemas; (3) the existence of abdominal infections.[1,5]

It seems that mucosal damage, as well as bowel distension secondary to increased bowel pressure in the setting of diagnostic studies, are the main factors that lead to the occurrence of HPVG in IBD patients.[1,2,4,5]

With the present case, only 18 cases of HPVG in patients with UC have been described in the English literature to date (Table 1).[1,6,7–21] Sixty one percent of cases were found after a barium enema or a colonoscopy. The majority of cases (88%) were managed conservatively. The mortality rate was 5% (one out of 18 patients), although it seems that in this case the fatal outcome was due to a complications occurred after an emergency surgery in the setting of fulminant colitis.[16]

Therefore, it seems that the management and treatment of HPVG has to be addressed to the underlying disease, which will determine the prognosis of the patients.

Author contributions

Murzi M, Oblitas E, Garcia-Planella E and Gordillo J designed the case report; Posso M provided advice on performing the systematic review and searched in the databases; Murzi M and Gordillo J screened the articles and selected the full texts; Murzi M wrote the paper; Soriano G, Gordillo J, Pernas JC and Garcia-Planella E provided clinical advice; Soriano G, Gordillo J and Garcia-Planella E revised the paper. All authors contributed and approved the last version of the manuscript.

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Conflict of interest

The authors declare that they have no conflict of interest.

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Esófago negro tras doble trasplante hepatorrenal

Black esophagus after combined liver-kidney transplantation

La necrosis esofágica aguda es una entidad infrecuente caracterizada por la presentación de pigmentación negra difusa del esófago en la endoscopia, de causa isquémica y que generalmente se manifiesta como hemorragia digestiva alta (HDA). La mortalidad es elevada y el manejo difiere entre tratamiento médico intensivo o cirugía cuando se asocia a perforación.

Se presenta un caso de esófago negro asociado a mediastinitis tras doble trasplante hepatorrenal (DTHR).

Se trataba de un varón de 50 años con diagnóstico de cirrosis hepática enólica Childh-Pugh A5, MELD 20, hipertensión portal e insuficiencia renal crónica en diálisis peritoneal. Se indicó DTHR. Durante la intervención precisó politransfusión y presentó una parada cardiaca con actividad eléctrica sin pulso durante 2 min. En el postoperatorio inmediato mantuvo estabilidad hemodinámica y las pruebas complementarias realizadas (analíticas, doppler seriados y colangiografía) fueron normales. En el decimotercer día postoperatorio presentó inestabilidad hemodinámica y HDA, realizándose una gastroscopia con hallazgo de esófago negro por necrosis circunferencial en toda su longitud. Se practicó una TC, donde se observó neumomediastino y una colección mediastínica (fig. 1A), indicándose intervención quirúrgica urgente. El esófago mostraba necrosis transmural con una perforación en el tercio inferior condicionando una mediastinitis. Se realizó esofaguectomía transhiatal, desbridamiento y lavado mediastínico, esofagostomía cervical y gastrostomía tipo Witzel. En el estudio anatomopatológico el esófago presentaba necrosis completa de la mucosa secundaria a esofagitis aguda necrosante (fig. 1B). La evolución fue lenta pero satisfactoria, presentando, al mes del trasplante, una trombosis postanastomótica de la arteria